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Parents' experiences of caring for a child with a cleft lip and/or palate: a review of the literature

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Abstract

This review brings together for the first time the existing quantitative and qualitative research evidence about the experiences of parents caring for a child with a cleft. It summarizes salient themes on the emotional, social and service-related experiences of parents and critiques the literature to date, comparing it with wider, selected literature from the field of children's long-term conditions, including disability. The review suggests that there are similarities and differences between the literatures, in terms of research focus and approach. Similarities are found across children's conditions in the perspectives of parents on emotional, social and service-related aspects, although much of the cleft literature is focused on the early stages of children's lives. However, the quality of cleft research to date about parents' experiences has also been variable, with a narrow emphasis on cross-sectional, deficit-orientated psychological approaches focused mainly on mothers. Despite a substantial literature, little qualitative research has examined parents' perspectives in-depth, particularly about their child's treatment journey. This contrasts with the wider children's literature, which has traditionally drawn not only on psychological approaches but also on the broader perspectives of sociology, social policy, nursing and health services research, using both qualitative and quantitative methods, often in integrated ways. Such approaches have been able to highlight a greater range of experiences from both mothers and fathers, about caring for a child with a long-term condition and views about treatment. The review identifies a lack of comparable research in the cleft field to examine parents' experiences and needs at different stages of their children's lives. Above all, research is needed to investigate how both mothers and fathers might experience the long-term and complex treatment journey as children become older and to elicit their views about decision making for cleft treatments, particularly elective surgeries.

Keywords
cleft lip, cleft palate, parents' experiences, parents' perceptions

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Introduction

Clefts of the lip and/or palate[1] are among the most commonly occurring congenital impairments (Mossey & Little 2002) and may affect a child's ability to feed, chew, breathe and hear, as well as cause disturbance to dental, facial, speech and language development (Clinical Standards Advisory Group 1998). Other possible consequences include effects on emotional and social well-being, behavioural and learning difficulties (Hunt *et al.* 2005). In the UK, a long-term programme of surgery, orthodontic treatment and speech therapy is offered by multi-disciplinary teams of specialist clinicians, to address the functional and appearance-related consequences of having a cleft.

[1] In this paper, the term 'cleft' refers collectively to clefts of the lip, clefts of the palate or clefts of both the lip and the palate.

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Table 1. Literature search strategy

Cleft literature search	
Databases searched: British Nursing Index CINAHL Plus EMBASE Health and Psychosocial Instruments Maternity and Infant Care MEDLINE PsychINFO Social Sciences Index Sociological Abstracts ISI Web of Knowledge **Hand searching:** Cleft Palate-Craniofacial Journal **Grey literature:** Unpublished papers, voluntary sector reports **Limitations:** from 1980 to present; human subjects; English language **Critical appraisal:** All studies appraised for quality using criteria for the critique of both qualitative research (Popay *et al*. 1998; Seale *et al*. 2004) and quantitative research [Greenhalgh 2001; Critical Appraisal Skills Programme (CASP) 2007]	**Search strategy** (combination of free text terms in title or abstract and Subject Headings): *Free text terms:* (parent* or mother* or father* or family or families) OR (child* or infant) OR (adolescen* or young people or young person* or teenage*) OR (paediatric* or pediatric*) AND (cleft lip or cleft palate or cleft lip and palate or craniofacial or cranio-facial) *Subject Headings:* British Nursing Index (CLEFT PALATE) AND (CHILDREN OR PARENTHOOD OR FAMILY) CINAHL Plus (CLEFT LIP OR CLEFT PALATE) AND (PARENTING OR PARENTS OR CHILD OR ADOLESCENT) EMBASE (CLEFT LIP OR CLEFT PALATE) AND (CHILD OR PARENT) Maternity and Infant Care (CLEFT LIP OR CLEFT PALATE) AND (CHILD HEALTH OR INFANT OR PARENTS) MEDLINE (CLEFT LIP OR CLEFT PALATE) AND (CHILD OR PARENTS)
Long-term conditions literature search	
Databases searched: British Nursing Index CINAHL Plus EMBASE Health and Psychosocial Instruments Maternity and Infant Care MEDLINE PsychINFO Social Sciences Index Sociological Abstracts ISI Web of Knowledge	**Search strategy** (free text terms in title or abstract): *Free text terms:* (parent* or mother* or father* or family or families) OR (child* or infant) OR (adolescen* or young people or young person* or teenage*) OR (paediatric* or pediatric*) AND (chronic disease or chronic illness) OR (long term condition or long-term condition) OR (disab*) OR (congenital) **Limitations:** from 1990 to present (because of changes in policy/theoretical perspectives since 1990s); human subjects; English language

Biomedical and psychological perspectives have predominated in cleft research to date (Rumsey & Harcourt 2005) and have focused mainly on the identification of risk factors and measurement of deficits (Eisermann 2001). Some research has also investigated people's views of service-related issues, although qualitative approaches to gauging in-depth perceptions have so far been relatively rare (Nelson 2009). This paper will present a narrative overview of the literature that examines the experiences of parents caring for a child with a cleft, making comparisons with wider, selected literature from the field of children's long-term conditions and disability. It will draw out key issues to highlight similarities and differences, as well as gaps in knowledge.

Literature search and appraisal

The literature was searched comprehensively to identify publications that have examined the experiences of parents of children with clefts and more selectively for articles about parents and long-term conditions including disability. The search strategy is presented in Table 1, while Table 2 presents a summary of the 57 publications found.

Emotional experiences of having a child with a cleft

Early experiences and needs

Discourses of 'loss', 'mourning' and 'correcting' have been common in research perspectives surrounding the diagnosis of a child with a cleft, informed by the assumptions of earlier theoretical perspectives (Olshansky 1962; Solnit & Stark 1962; Drotar *et al*. 1975). Both pre- and post-natally, across countries and cultures, parents' feelings of shock, anger, grief and worry have been documented both in surveys and in qualitative studies (Bradbury & Hewison 1994; Rey-Bellet & Hohlfeld

Table 2. Details of cleft lip and palate studies included in the review

Author and year	Focus of the study	Methods	Sample size (parents)	Strengths and limitations
Mixed-methods studies				
Benson *et al.* 1991	*Coping/adaptation (social support)* USA	Questionnaires Semi-structured interviews	72	Matched comparison child group; validated measure (Social Support Questionnaire – Revised) Response rate/parent characteristics missing; fathers under-represented
Bradbury and Hewison 1994	*Coping/adaptation* UK	Semi-structured interviews Questionnaire	59	Recruitment details/sample characteristics missing; qualitative data collection/analysis not fully described
Cadogan *et al.* 2009	*Information (diagnosis)* UK	Questionnaire incorporating both closed and open questions	31	100% response rate; longitudinal Small sample; qualitative analysis details missing
Cleft Lip and Palate Association 2007 *Voluntary sector report*	*Services (care provision)* UK	Questionnaire with some open-ended questions	227	40% response rate Sampling strategy/characteristics missing and mainly from parent support group
Eisermann 2001	*Positive outcomes* USA	Questionnaire Unstructured interviews	11	Comparable/contrasting cases addressed Response rate missing; convenience sampling; sample not diverse and mainly from support organizations; gender mix of sample missing
Martin 1995	*Information (diagnosis)* UK	Questionnaire	41	Fathers and mothers; 70% response rate Sample characteristics/instrument details/children's ages missing; sample from support group only; qualitative analysis missing
Nelson *et al.* 2009	*Perceptions of cause* UK	Questionnaire incorporating open-ended items	42	Some validated scales (Hospital Anxiety and Depression Scale, Perceived Stress Scale) Framework analysis 21% response rate
Pelchat *et al.* 2004	*Coping/adaptation (support from services)* Canada	Questionnaire and free comments to each question	76	80% response rate Analysis of qualitative data not described
Slade *et al.* 1999	*Coping/adaptation* UK	Questionnaires Structured interviews	32	88% response rate; validated instruments (Symptom Checklist 90-R, Edinburgh Postnatal Depression Scale, Self Perception Inventory, Neonatal Perception Inventory, Impact of Event Scale, Parental Competence Scale); longitudinal Small sample size; qualitative analysis not fully described
Qualitative studies				
Cartwright and Magee 2006 *Voluntary sector report*	*Information (needs)* UK	Focus groups One-to-one interviews	3	In-depth focus; framework analysis Response rate missing; sample small and from support group only
Chuacharoen *et al.* 2009	*Information (needs)* Thailand	Semi-structured interviews	15	Longitudinal; independent interviewer; content analysis Response rate/recruitment details/sampling strategy missing
Farrimond and Morris 2004 *Unpublished undergraduate dissertation*	*Information (diagnosis)* UK	Semi-structured interviews	10	In-depth/flexible approach; reflexive stance; interpretive Phenomenological analysis Response rate missing
Johansson and Ringsberg 2004	*Coping/adaptation (social support)* Sweden	Semi-structured interviews	30	In-depth focus; use of phenomenology Response rate missing; sample not diverse
Klein *et al.* 2006	*Parenting* USA	Semi-structured interviews	9	In-depth focus Response rate missing; sample from parent support organization only
Martin 2005	*Information (prenatal diagnosis)* UK	Structured interviews	10	100% response rate; mothers and fathers Ages of children missing; little flexibility in study instrument; analysis techniques/reflexive stance missing

Table 2. *Continued*

Author and year	Focus of the study	Methods	Sample size (parents)	Strengths and limitations
Nusbaum *et al*. 2008	*Information (diagnosis)* USA	Semi-structured interviews	20	63% response rate; in-depth/flexible approach; includes comparable and contrasting findings; reflexive stance Sample lacks variation
Owens 2008	*Services (feeding support)* UK	Narrative interviews	20	Recruitment/sample details missing; few details of analysis techniques; no treatment of negative cases
Quantitative studies				
Andrews-Casal *et al*. 1998	*Coping/adaptation* USA	Questionnaires	61	52% response rate; some validated instruments (Parenting Stress Index, Hollingshead Index of Social Position) Gender mix of sample unclear
Baker *et al*. 2009	*Coping/adaptation* UK	Questionnaires	103	Validated study instruments (Coping Response Inventory, Interpersonal Support Evaluation List-Short Form, Stress-Related Growth Scale, General Health Questionnaire, Family Impact Scale) Response rate unclear; fathers under-represented
Barden *et al*. 1989	*Mother–infant interaction/ attachment* USA	Observation Questionnaires	10	Comparison group Recruitment/response rate details missing; small and diverse sample; laboratory conditions; cross-sectional
Berger and Dalton 2009	*Coping/adaptation* UK	Questionnaire	143	Reasonable sample size (multi-centre); validated instruments (General Well-Being Scale, Brief COPE, Satisfaction with Appearance Questionnaire) 37% response rate; unrepresentative sample; fathers under-represented
Black *et al*. 2009	*Adaptation* Thailand, China, Colombia	Questionnaire	98	Sample size reasonable and representative Recruitment details/children's ages missing; no fathers
Broder *et al*. 1992	*Services (outcome of care)* USA	Standardized interviews	495	Good sample size Recruitment/sample details/response rate unclear
Broder and Trier 1985	*Information (diagnosis)* USA	Questionnaire	37	46% response rate; sample small and characteristics missing
Byrnes *et al*. 2003	*Information (diagnosis)* USA	Questionnaire	98	Reasonable sample size 43% response rate; fathers under-represented
Campis *et al*. 1995	*Coping/adaptation* USA	Questionnaires	77	95% response rate; validated instruments (Child Behavior Checklist, Beck Depression Inventory, Spielberger Trait Anxiety Scale, Parenting Stress Index, Social Support Questionnaire – Revised) Sample unrepresentative
Canady *et al*. 1997	*Services (continuity of care)* USA	Questionnaire	96	73% response rate; few sample or study instrument details
Cleft Lip and Palate Association 1996 *Voluntary sector report*	*Services (care provision)* UK-wide	Questionnaire	102	100% response rate Sampling strategy/characteristics missing; sample from parent support group only
Davalbhakta and Hall 2000	*Information (diagnosis)* UK	Questionnaire	90	78% response rate; sample size reasonable Sample characteristics/instrument details missing
Endriga and Speltz 1997	*Mother–infant interaction and attachment* USA	Observation	116	Matched control group Response rate missing; cross-sectional; laboratory-based

Table 2. *Continued*

Author and year	Focus of the study	Methods	Sample size (parents)	Strengths and limitations
Field and Vega-Lahr 1984	*Mother–infant interaction and attachment* USA	Observation	24	Recruitment/response rate missing; small sample; control group matched on some variables but not fully described
Jeffery and Boorman 2001	*Services (care provision)* UK	Questionnaire	341	72% response rate; good sample size Sample characteristics missing
Kramer *et al*. 2007	*Quality of life* Germany	Questionnaire	130	84% response rate; validated instrument (Impact on Family Scale) Gender mix of sample unclear
Krueckeberg and Kapp-Simon 1993	*Coping adaptation* USA	Questionnaires	52	Comparison group; validated instruments (Parenting Stress Index, Modification of the Block Child Rearing Practices Report, Social Relationship Scale, Four Factor Index of Social Status, Social Skills Questionnaire) Response rate missing; sample relatively small; gender mix of sample missing
Locker *et al*. 2002	*Quality of life* Canada	Questionnaire	93	Validated scale (Family Impact Scale) Recruitment/response rate and sample characteristics missing
Murray *et al*. 2008	*Mother–infant interaction and attachment* UK	Observation Questionnaires	190	75% response rate; control group; home environment; some validated instruments (Behaviour Screening Questionnaire, Structured Clinical Interview for DSM Diagnoses)
Noar 1991	*Services (outcome of care)* UK	Questionnaire	30	Instrument piloted Recruitment/response rate and sample details missing; small sample
Noor and Musa 2007	*Services (care provision/ outcome)* Malaysia	Standardized interview	60	Validated instrument (Cleft Evaluation Profile) Recruitment/response rate details missing; gender mix of sample unclear
Oliver and Jones 1997	*Services (feeding support)* UK	Questionnaire	100	64% response rate Children's ages/sample characteristics/study instrument details missing
Pannbacker and Scheuerle 1993	*Decision making* USA	Questionnaire	42	56% response rate Small sample; children's ages/sample characteristics/study instrument details missing
Pelchat *et al*. 1999	*Coping/adaptation* Canada	Questionnaires	74	91% response rate; matched comparison groups; validated instruments (Stress Appraisal Measure, Parenting Stress Index, Emotional Distress Index-Quebec Health and Social Survey); longitudinal
Pope *et al*. 2005	*Coping/adaptation* USA	Questionnaires	47	Longitudinal, validated instruments (Parenting Stress Index-Short Form, Child Behavior Checklist) Response rate unclear; sample characteristics and gender mix unclear
Rey-Bellet and Hohlfeld 2004	*Information/ counselling* Switzerland	Questionnaire	29	82% response rate Parents' socio-economic characteristics missing; analysis details missing; unclear how many fathers participated
Sank *et al*. 2003	*Coping/adaptation (social support)* USA	Questionnaires	145	98% response rate; validated instruments (Interpersonal Support Evaluation List, Beck Depression Inventory) No fathers
Semb *et al*. 2005	*Services (care provision/ outcome)* Denmark, Norway, UK, Netherlands, Sweden	Questionnaire	81	65% response rate; questionnaire piloted; longitudinal Sample characteristics of parents missing

Table 2. *Continued*

Author and year	Focus of the study	Methods	Sample size (parents)	Strengths and limitations
Speltz *et al*. 1990	*Coping/adaptation* USA	Questionnaires Observation	55	Matched control group; validated instruments (Parenting Stress Index, General Well-Being Scale, Social Health Battery, Locke-Wallace Marital Adjustment Scale) Response rate missing; small sample
Speltz *et al*. 1993	*Coping/adaptation* USA	Questionnaires	33	Control group; validated instruments (Dyadic Parent-Child Interaction Coding System, Nursing Child Assessment Teaching Scale, General Well-Being Schedule, Social Network Reciprocity and Dimensionality Assessment Tool, Dyadic Adjustment Scale, Child Behavior Checklist); longitudinal Small sample
Speltz *et al*. 1994	*Mother–infant interaction and attachment* USA	Questionnaires Observation	51	100% response rate; control group Clinic conditions; cross-sectional
Speltz *et al*. 1997	*Mother–infant interaction and attachment* USA	Questionnaires	115	Control group; validated instruments (Mental Health Index, Parenting Stress Index, Dyadic Adjustment Scale, Family Environment Scale, Questionnaire on Social Support); longitudinal design Response rate missing; laboratory-based
Strauss *et al*. 1988	*Services (outcome of care)* USA	Standardized interviews	102	Study instrument piloted Recruitment/response rate and parent sample characteristics missing
Strauss *et al*. 1995	*Information (diagnosis)* USA	Questionnaire	100	76% response rate; reasonable sample size; survey instrument piloted Fathers under-represented
Strauss *et al*. 2007	*Experiences of stigmatization* USA	Questionnaire	153	Reasonable sample size; response rate missing; convenience sample; fathers excluded
Thomas *et al*. 1997	*Services (outcome of care)* UK	Questionnaire	62	70% response rate Parent sample characteristics missing
Turner *et al*. 1997	*Services (care provision/ outcome)* UK	Standardized interview	130	76% response rate Recruitment/parent sample details missing
Turner and Milward 1988	*Services (care provision)* UK	Structured interviews	60	Recruitment/response rate/details of study instrument missing; gender mix of sample unclear
Williams *et al*. 2001	*Services (care provision/ outcome)* UK	Questionnaire	438	76% response rate; robust sample size; multi-centre Parent sample characteristics missing
Young *et al*. 2001	*Information (diagnosis)* USA	Questionnaire	40	100% response rate Sample unrepresentative; fathers under-represented

2004; Martin 2005; Black *et al*. 2009; Cadogan *et al*. 2009; Chuacharoen *et al*. 2009). In-depth qualitative research, however, has been able to reveal a wider range of experience, often highlighting parents' elation at a child's birth, or perceptions of a child's cleft as unremarkable or unique (Eisermann 2001; Farrimond & Morris 2004; Johansson & Ringsberg 2004; Klein *et al*. 2006). Feelings of parental guilt, self-blame and associated anxiety have also been described (Strauss *et al*. 1995; Byrnes *et al*. 2003; Nelson *et al*. 2009) with recognition that parents may to wish share their feelings and get emotional support from experienced professionals at the time of diagnosis (Martin 1995; Strauss *et al*. 1995; Byrnes *et al*. 2003; Johansson & Ringsberg 2004;

Rey-Bellet & Hohlfeld 2004). The quality of both quantitative and qualitative studies in this body of research is variable, however, with half omitting to provide response rates and a quarter to document children's ages. Most exclude fathers and focus on the collection of cross-sectional data alone. In addition, the quantitative studies comprise samples as small as 25 and use few validated measures, while little of the qualitative research fully presents its approach or findings.

Studies of the early developing relationship between parents and children with clefts have been strongly informed by attachment theory (Bowlby 1965) and suggest that infants may be as securely attached to their mothers as those without clefts (Speltz *et al.* 1990, 1993, 1997; Endriga & Speltz 1997; Slade *et al.* 1999), with a potentially less sensitive interplay between the two because of possible disturbances in communication cues (Field & Vega-Lahr 1984; Barden *et al.* 1989; Speltz *et al.* 1990, 1994; Endriga & Speltz 1997; Murray *et al.* 2008). Although the majority have used validated instruments, their findings are based on relatively small and diverse samples, comprised of between $n = 10$ and $n = 55$. Only four took a longitudinal approach to track developments over time, but their groups were inconsistently matched, sometimes including children with associated developmental difficulties as well as a cleft (Speltz *et al.* 1993, 1997; Murray *et al.* 2008). Most notably, only one study (Murray *et al.* 2008), which was also the most robust in terms of sample size at $n = 190$, observed mothers and infants in their own homes, the remainder having been conducted in artificial laboratory conditions. Attachment theory has been widely criticized outside the field on the grounds that it takes no account of social or environmental context on the developing parent–child relationship (Knestrict 2002) and reinforces stereotypical maternal roles, attributing responsibility for a child's emotional and social difficulties to women alone (Contratto 2002).

Although feeding difficulties have been reported among parents of infants with clefts in one UK survey ($n = 100$; Oliver & Jones 1997), research to explore parents' emotional experiences of feeding their child has been relatively rare. Only one qualitative study has offered some insight into the emotional impact on mothers of pressure to successfully breastfeed (Owens 2008), but it provides little information on its sampling and recruitment strategy or details of whether any cases might have contrasted with the analysis presented.

Emotional 'strain'

Several psychological studies based in the USA have focused on the assessment of stress levels among parents caring for children with clefts, investigating outcomes such as anxiety, depression and poor psychological 'adjustment', collectively referred to here as emotional 'strain'. Some studies have found evidence of raised levels of emotional strain among parents in their child's toddlerhood (Speltz *et al.* 1990, 1993; Pope *et al.* 2005), but findings generally suggest that by pre-school years, levels are equivalent to those in parents of children without clefts (Krueckeberg & Kapp-Simon 1993; Campis *et al.* 1995; Andrews-Casal *et al.* 1998; Pelchat *et al.* 1999; Slade *et al.* 1999; Berger & Dalton 2009). Most of these studies have used relatively small sample sizes (average $n = 58$) along with diverse research instruments, making inferences difficult to draw. Further, accounts from parents in their own words are rare and the focus of research has primarily been on the negative experiences associated with caring for a child with a cleft, to the relative exclusion of potentially positive aspects (Eisermann 2001; Baker *et al.* 2009).

Some studies have suggested that a child's cleft treatment and clinic attendances may impact adversely on families' quality of life (Locker *et al.* 2002; Baker *et al.* 2009), and outside the UK a small number of in-depth qualitative studies have highlighted parents' feelings of anxiety about surgery and their perceived need for emotional support through treatment for both themselves and their child (Turner *et al.* 1997; Eisermann 2001; Johansson & Ringsberg 2004; Klein *et al.* 2006). However, as far as is known, no previous studies have aimed to explore the emotional experiences of parents in connection with their child's treatment pathway.

Coping

Research in the field on parental coping is subsumed in the studies outlined in the preceding section; however, it is important to highlight that the focus of this work has been mainly on the cross-sectional assessment of poor 'adjustment' in deficit-oriented ways (Baker *et al.* 2009) with no longitudinal work to track parental coping as a child becomes older. Contextual factors have largely been excluded in this body of work too, with studies taking little account of issues other than a child's cleft that may be affecting families' lives (Krueckeberg & Kapp-Simon 1993). The incorporation of 'resilience' models in the investigation of coping in cleft research has been highlighted as a desirable goal (Broder 2001; Kapp-Simon & Gaither 2009), yet little published research of this kind exists to date. However, influenced by models of resilience and positive coping, a recent British study ($n = 103$), using validated study instruments including the Coping Response Inventory and the Stress Related Growth Scale, investigated coping strategies and

perceptions of positive outcomes among parents (Baker *et al.* 2009). The study found a high degree of positive coping and outlook, particularly in parents of children with more extensive clefts.

Studies using mainly qualitative methods have also offered insights into the positive and/or rewarding aspects of caring for a child with a cleft that have been identified by parents themselves. Such rewards include recognition of their own personal strengths, stronger relationships, appreciation of diversity and others' good intentions, tolerance, a sense of community and hope for the future (Bradbury & Hewison 1994; Eisermann 2001; Klein *et al.* 2006). Parents have also reported recognizing their child's strengths, such as a determined attitude, perseverance and sociability (Klein *et al.* 2006). In one study, half of parents reported that they would not remove their child's cleft if given the opportunity (Eisermann 2001).

In terms of coping resources, it has been suggested that because of the stigma potentially associated with having a visible facial difference, parents of children with clefts may lack adequate social support (Benson *et al.* 1991; Speltz *et al.* 1993; Pelchat *et al.* 1999; Sank *et al.* 2003). Some studies have found low levels of social support among such parents (Benson *et al.* 1991; Speltz *et al.* 1993; Campis *et al.* 1995; Sank *et al.* 2003), while others find no particular differences between parents of children with and without clefts (Krueckeberg & Kapp-Simon 1993). Yet others find variation across families, regardless of their child having a cleft (Bradbury & Hewison 1994; Johansson & Ringsberg 2004; Baker *et al.* 2009). However, these studies have not clearly delineated different types of social support and how it might be experienced by parents, distinguishing, for example, the relative impact of formal and informal support.

Investigation of the coping strategies used by parents does not appear to have been a priority in cleft research to date, although a small amount of qualitative work has indirectly revealed some of the strategies which parents may use. These include cognitive or problem-focused strategies such as discussing a child's needs with school staff and thinking of ways to help them establish friendships/handle teasing (Klein *et al.* 2006). Parents have also reported using emotion-focused strategies such as maintaining a hopeful attitude for the future and a belief in their own competence as parents (Johansson & Ringsberg 2004; Klein *et al.* 2006).

Social experiences related to having a child with a cleft

A smaller body of research has provided insights into parents' social experiences. It is known that children and their families may be 'stigmatized' because of a cleft's effects on appearance and/or function (Goffman 1963; Partridge 1997) and their social experiences characterized by discomfort, anxiety or rejection because of perceived 'differences' (Rumsey & Harcourt 2005). Research which has been mainly qualitative in nature has revealed the challenges that parents may face in managing others' reactions to their child's cleft. Mothers have described their heightened sensitivity to the reactions of friends and family (Farrimond & Morris 2004; Johansson & Ringsberg 2004), the verbal and non-verbal expressions of distaste encountered in public and attempts to conceal their baby or shun social situations in order to avoid feeling 'different' (Bradbury & Hewison 1994; Johansson & Ringsberg 2004; Klein *et al.* 2006).

Parents have also reported in qualitative studies their worries about social issues, including concerns about a child's acceptance by peers, experiences of teasing, finding a life partner and securing employment (Cartwright & Magee 2006; Klein *et al.* 2006). Klein and colleagues' in-depth study recorded the distress caused to parents by their child's reported experiences of teasing or bullying at school, or when entering new and unfamiliar settings. This study also uncovered the tension parents experienced in efforts to protect children, while promoting their independence. One survey from the USA (n = 153), reported perceived stigmatizing experiences to be prevalent among both mothers and their children (Strauss *et al.* 2007). In a number of others across countries, between 15% and 68% of parents reported feeling their child's self-confidence to have been affected by having a cleft and between 50% and 68% felt their child had been teased (Noar 1991; Turner *et al.* 1997; Semb *et al.* 2005; Noor & Musa 2007). Overall, this research has been more balanced in terms of its methods, with quantitative surveys and qualitative studies more equally employed, although fathers' perspectives remain rare.

Experiences of cleft services

Information and decision making

The majority of cleft studies have focused on parents' information experiences and needs at the point of diagnosis, with fewer studies addressing these issues across children's age ranges. However, a lack of readily accessible information at diagnosis has been reported (Martin 1995; Cleft Lip and Palate Association 1996; Young *et al.* 2001), as has parents' need for accurate and balanced information about clefts and their causes (Davalbhakta & Hall 2000; Young *et al.* 2001; Nusbaum *et al.* 2008). Studies have also suggested that most parents prefer to receive

their information at this time verbally, from specialist practitioners (Strauss *et al.* 1995; Byrnes *et al.* 2003).

While the involvement of parents and children in decisions about their care has been encouraged in UK health policy, research about parents' experiences of decision making for their child's cleft treatment is sparse. One small survey from the USA ($n = 42$) found that over a third of parents wanted to be more involved in decision making about treatment (Pannbacker & Scheuerle 1993), and surveys in the UK have also suggested that some parents do not feel involved in decisions and would like more involvement (Turner *et al.* 1997; Jeffery & Boorman 2001).

Experiences of service organization, delivery and outcomes

'Satisfaction' with the organization and delivery of cleft services has been investigated in a number of surveys whose results suggest high ratings among parents both in the UK and elsewhere (Turner *et al.* 1997; Jeffery & Boorman 2001; Williams *et al.* 2001; Semb *et al.* 2005; Cleft Lip and Palate Association 2007; Kramer *et al.* 2007; Noor & Musa 2007). Satisfaction has often been treated rather simplistically in this research, however, as most papers fail to properly define how it is being conceptualized. Some refer to satisfaction with 'care and attention' (Turner *et al.* 1997; Williams *et al.* 2001; Noor & Musa 2007), some to the 'level' of care (Jeffery & Boorman 2001) and others to the 'manner' of care provided (Semb *et al.* 2005). Despite the positive ratings of parents reported, areas of concern have also been identified, such as poor access to and co-ordination of services (Martin 1995; Cleft Lip and Palate Association 1996, 2007; Oliver & Jones 1997). However, these surveys comprise widely differing sample sizes (between $n = 30$ and $n = 495$) and are almost exclusively cross-sectional and apart from the largest study (Williams *et al.* 2001), from single centres. Further, study instruments in this body of research are diverse and validated measures largely absent.

Research suggests that parents have confidence in cleft clinicians because of their specialism (Johansson & Ringsberg 2004; Semb *et al.* 2005; Cleft Lip and Palate Association 2007) and that they have concerns about the lack of knowledge among generic healthcare staff (Turner & Milward 1988; Cleft Lip and Palate Association 1996; Oliver & Jones 1997; Johansson & Ringsberg 2004; Martin 2005; Cartwright & Magee 2006). Parents have highlighted the importance of having practitioners who communicate well and show sensitivity (Broder & Trier 1985; Cleft Lip and Palate Association 1996, 2007; Jeffery & Boorman 2001; Byrnes *et al.* 2003; Semb *et al.* 2005; Cartwright & Magee 2006) and have identified continuity of care from specialist cleft clinicians as especially valuable (Cleft Lip and Palate Association 1996; Canady *et al.* 1997).

Some literature has also focused on parents' satisfaction with the outcomes of their children's cleft treatment, using the key variables of facial appearance, function and psychosocial well-being. These studies too have commonly reported high ratings of satisfaction with treatment (Strauss *et al.* 1988; Noar 1991; Broder *et al.* 1992; Thomas *et al.* 1997; Turner *et al.* 1997; Williams *et al.* 2001; Pelchat *et al.* 2004; Semb *et al.* 2005; Noor & Musa 2007; Berger & Dalton 2009). Some have also found broad agreement between parents and their children (Strauss *et al.* 1988; Semb *et al.* 2005; Noor & Musa 2007), while others have revealed dissimilar views on different variables (Noar 1991; Broder *et al.* 1992; Thomas *et al.* 1997; Williams *et al.* 2001; Berger & Dalton 2009). On outcomes for psychosocial well-being, findings have also been inconsistent, with some suggesting children with clefts to experience low levels of self-confidence and high levels of teasing (Semb *et al.* 2005; Noor & Musa 2007). Others report children to be less emotionally and socially affected by their cleft than their parents estimate (Noar 1991), or to experience more social, but fewer emotional effects (Turner *et al.* 1997). Differences in these findings may be due to the diverse samples and study instruments used, a lack of longitudinal data or multidimensional approaches to researching satisfaction with treatment results in a particularly sensitive context, or differing personal/cultural expectations about the outcomes of care.

Discussion

This literature review has contributed knowledge about the emotional, social and service-related experiences of parents caring for a child with a cleft, but has highlighted the variable quality of research to date. It has demonstrated that quantitative research in the cleft field has comprised mainly cross-sectional surveys, with relatively small sample sizes focused mainly on mothers. Little qualitative research, either standing alone or as part of mixed-methods designs, has been carried out to investigate parents' perceptions and experiences and varies widely in the extent to which it takes an in-depth approach. Similarities as well as differences are found in the wider literature on children's long-term conditions.

Comparison and contrast with the wider literature

Parents may encounter particular emotional and social challenges because of a cleft's visible and/or audible effects on

their child's facial appearance and speech – both of central importance to interpersonal relationships and communication (Rumsey & Harcourt 2005). None the less, similarities have been found among the parents of children with other long-term conditions in relation to their emotional, social and service-related experiences. Recognition of the potential emotional impact of a child's diagnosis and the need for adequate early emotional and social support is comparable (Grootenhuis & Last 1997; Sloper 1999; Case 2000; Piggot *et al.* 2002; Trulsson & Klingberg 2003; Barr & McConkey 2007). Parental experiences of social stigma are also reflected in the long-term conditions literature, as is the tension parents may experience between protecting children while fostering their independence (Ray 2003; Rehm & Bradley 2005; Duguid *et al.* 2007; Lassetter *et al.* 2007). Also found are parents' concerns about accessible, well-coordinated services for children (Mitchell & Sloper 2001; Watson *et al.* 2002; Law *et al.* 2003; Beecham *et al.* 2007) and needs for information (Mitchell & Sloper 2002; Lawoko 2007). A need for knowledgeable, sensitive practitioners with good communication skills is also reflected here (Davies *et al.* 2003; Farrant & Watson 2004; Hallstrom & Elander 2007), as is the value placed on long-term relationships between families and practitioners (Trulsson & Klingberg 2003; Lalor *et al.* 2007). Finally, in common with the cleft literature, high levels of parent satisfaction with treatment outcome are reported in studies of children's surgery (Bridwell *et al.* 1999; Smith *et al.* 2006) with similar inconsistencies of opinion between parents and children (Bridwell *et al.* 1999; Pratt *et al.* 2002; Rinella *et al.* 2004; Smith *et al.* 2006).

Marked differences can be found in the children's long-term conditions literature, however, in terms of both research approaches and focus, particularly in relation to the emotional and service-related aspects of parents' experiences. This literature draws on broader perspectives from sociology, social policy, nursing and health services research, more commonly recognizing the value of both quantitative and qualitative methods to study parents' experiences, often in integrated ways (Lassetter *et al.* 2007). Observation of naturally occurring interactions between parents and their children in everyday life and interviews to explore both mothers' and fathers' perceptions of the evolving nature of relationships over time are found (Anderson 1981; Lauritzen 1997; Kirk *et al.* 2005; Santos & McCollum 2007). The importance of the role of expectations and the need for multiple approaches to investigating parents' satisfaction with services, including in-depth qualitative studies, has also been emphasized here (Callery & Luker 1996; Smith *et al.* 2006; Green *et al.* 2008).

The variation in parents' reactions has been more widely acknowledged outside the cleft field in relation to diagnosis (Quine & Pahl 1987; Lane Tanner *et al.* 1998; Case 2000; Vehkakoski 2007; Bainbridge 2009), and the ambiguity of parents' feelings has been more evenly described (Larson 1998; Kearney & Griffen 2001; Nelson 2002; Carnevale *et al.* 2006). The ability of parents to cope and adapt is highlighted in the wider literature, informed by a change in perspective in the 1990s from pathologizing approaches to 'ecological' and 'resilience' models (McCubbin & McCubbin 1993; Beresford 1994; Wallander & Varni 1995; Sloper 1999; Rolland & Walsh 2006). Conceptual frameworks have documented that parents caring for disabled children through treatment may use a wide range of coping strategies, both problem- and emotion-focused (Burr & Klein 1994; Graungaard & Skov 2006), and the rewards of caring have also been more widely recognized, with evidence of parents' personal satisfaction and growth (Gregory 1994; Kearney & Griffen 2001; Nelson 2002; Barnett *et al.* 2006; King *et al.* 2006; Lassetter *et al.* 2007). Although rarely seen in the cleft literature, the emotional impact on parents of children's surgery has been a focus of research in the wider literature (Sobo 2005; Amin *et al.* 2006; Ben-Amitay *et al.* 2006; Joseph *et al.* 2007; MacLaren & Kain 2008), and parents' views of their children's treatment outcomes as often uncertain have also been brought to light (Lane Tanner *et al.* 1998; Kearney & Griffen 2001; Brinchmann *et al.* 2002; Vehkakoski 2007).

In contrast to cleft research, the wider literature draws heavily on the notion of flexible, holistic services to support families in different ways according to their needs as they move through treatment (McConachie 1994; King *et al.* 1997; Mitchell & Sloper 2001; Watson *et al.* 2002). Theories of 'respectful' or 'family-centred' care have long underpinned research on quality in family services in the wider children's literature (Mittler 1994; Mitchell & Sloper 2001; Trulsson & Klingberg 2003) but are relatively rare in the cleft literature so far. Additionally, theoretical perspectives about family involvement in decision making for treatment comprising ideas of 'partnership' between parents and practitioners (Coyne 1997; King *et al.* 1997; Piggot *et al.* 2002; Coyne & Cowley 2007) and family 'empowerment' (Mittler 1994; Mitchell & Sloper 2001) are central to the wider literature, but absent in cleft research. A substantial body of work in the wider children's literature has also investigated the experiences and preferences of parents about involvement in decision making about their child's care, using a range of research methods (Ellis & Leventhal 1993; Pyke-Grimm *et al.* 1999; Brinchmann *et al.* 2002; Hallstrom *et al.* 2002; Hallstrom & Elander 2004; Knopf *et al.* 2008). This research suggests that preferences cannot be predicted on the basis of demographics alone, but depend on

the context and nature of particular decisions (Knopf *et al.* 2008).

Conclusions

This review is the first to bring together evidence from both quantitative and qualitative research about the experiences of parents caring for a child with a cleft and demonstrates the variable quality of research to date. It highlights that despite a substantial literature, little research has so far examined parents' perspectives in-depth, with a narrow emphasis on cross-sectional, deficit-orientated psychological approaches focused mainly on mothers. Several gaps have been identified, including the lack of research to examine parents' experiences and needs at different stages of their children's lives, as they move through cleft treatment. Above all, research is needed to investigate how both mothers and fathers might experience their child's long-term and complex treatment journey as children become older and to elicit their views about decision making for cleft treatments, particularly elective surgeries.

Key messages

- This review synthesizes evidence from both quantitative and qualitative research about the experiences of parents caring for a child with a cleft.
- The quality of research on this topic to date has been variable.
- There has been a narrow emphasis on cross-sectional, deficit-orientated psychological approaches focused mainly on mothers.
- Despite a substantial literature, little qualitative research has examined parents' perspectives in-depth across children's ages.
- There has been a lack of research to examine in particular the experiences of mothers and fathers along their child's long-term and complex treatment journey including their views about decision making for cleft treatments.

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